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Prenatal sonographic diagnosis of congenital sacrococcygeal teratoma and management

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1 Introduction

Teratomas are tumors containing tissues derived from all three germinal cell layers, i.e.: ectoderm, endoderm, and mesoderm. Most teratomas in infancy and childhood arise in the sacrococcygeal region, with a reported incidence of 1/40.000 births. They are usually skin-covered masses protruding from the sacral area, causing distortion of the perineum and the buttocks, being three or four times more frequent in females, and about 10% are malignant [5, 16, 18]. During pregnancy, sacrococcygeal teratomas have been associated with both acute and chronic hydramnios, placentomegaly, and hydrops fetalis [2, 3, 8, 11]. During labor, severe dystocia may occur in 6% to 12% of the cases, and this is usually encountered in cases of infants with large tumors delivered vaginally [4, 13, 15, 19]. Reported is a case of a large sacrococcygeal teratoma diagnosed antepartum by sonography, and the perinatal management is briefly discussed.

2 Case report

C. V. S., a 25-year-old woman, gravida 3, para 2, was referred at 36 weeks' menstrual age due to discrepancy between dates and uterine size. No other complications during current pregnancy were found. Sonographic evaluation revealed a single fetus in vertex presentation, with a biparietal diameter and femur length compatible with her last menstrual period. A 15-cm by 12-cm sacral complex mass with predominant solid components was visualized (figure 1 A, B). Amniotic fluid volume was normal, and no associated anomalies were found. A sonographic diagnosis of sacrococcygeal teratoma was made. The patient refused hospitalization for further studies, and was admitted with spontaneous labor at 40 weeks' gestation. In view of the prenatal sonographic finding, an obstructed delivery could be anticipated. An emergency cesarean section was performed, delivering a 4.050-g female infant with a large sacrococcygeal tumor (figure 2), with Apgar score of 9 at 1 and 5 min. Some difficulty was found in delivering the buttocks, but this was easy accomplished by performing an inverted T-shaped hysterotomy. The fetal sacral mass was resected on the 5th day of life. The excised mass weighed 620-g, measured 14-cm by 11-cm by 9-cm, and the pathology reported a mature sacrococcygeal teratoma (figure 3). The infant had an uneventful recovery.

Curriculum vitae

WALDO H. SEPÚLVEDA, *M. D., was born in 1957 in Rancagua, Chile. He studied medicine at the University of Chile from 1975 to 1981, and was graduated in January 5, 1982. He specialized in obstetrics and gynecology at the University of Concepción, Chile, from 1982 to 1985. He is currently teaching and working at the University of Concepción, and his main fields of interest are ultrasonography, embryology, and perinatology.*

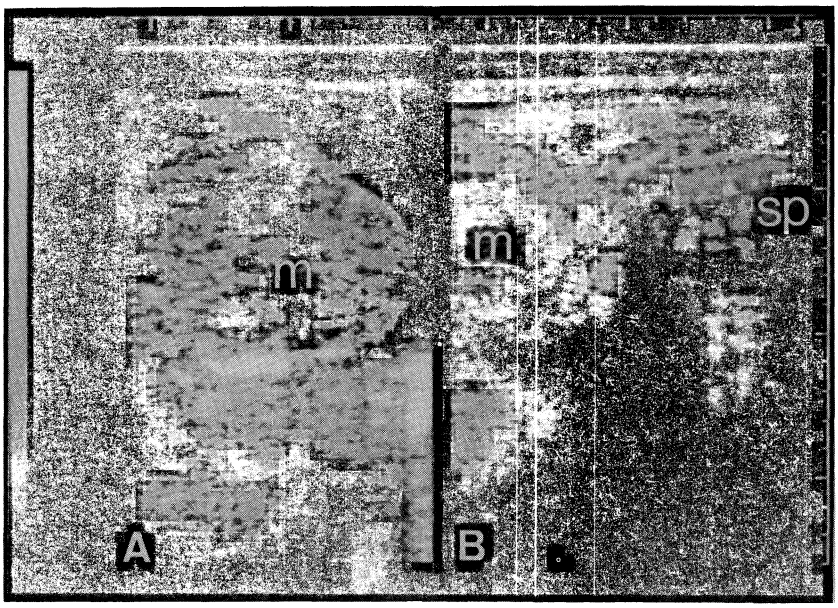

Figure 1. A. Antenatal sonogram showing a predominantly solid complex mass (m) with irregular cystic areas. **B.** Longitudinal scan showing the mass (m) attached to the fetal rump, unrelated to the fetal spine (sp).

Figure 2. Female newborn infant with a large teratoma protruding from the sacrococcygeal region.

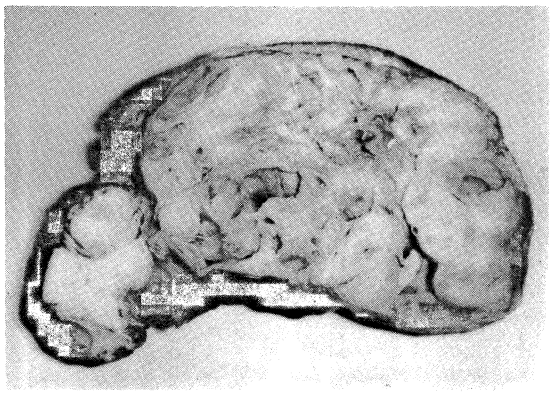

Figure 3. Mature sacrococcygeal teratoma.

3 Discussion

The classical clinical feature of a congenital sacrococcygeal teratoma is an obstructed delivery at the level of the fetal lower thorax after the head and shoulders are delivered without difficulty. Management in this situation includes vaginal incisional drainage [19] or blind dissection of the tumor [1], vaginal delivery after abdominal drainage of the tumor [4, 15], and emergency abdominal

delivery after partial vaginal delivery [13]. Often, fetal morbidity and mortality associated with this type of dystocia is extremely high.

The cornerstone in the management of sacrococcygeal teratomas is prenatal diagnosis. With the more frequent use of sonography, congenital sacrococcygeal teratomas are increasingly diagnosed antepartum [2, 6, 7, 9, 10, 14, 17]. Once the diagnosis is made, serial sonographic examinations have been advocated [2, 9, 17]. Sonography allows not only measurements of the tumor, but also its characterization as to whether it is predominantly solid or cystic, its documentation as to growth or changes in the tumor itself during pregnancy, and facilitates interventional procedures such as transabdominal puncture prior to cesarean section [12].

Some controversy exists regarding the critical size of the tumor and the choice of vaginal or abdominal delivery. Teal et al. [17] considered that an infant with a sacrococcygeal teratoma measuring more than 10-cm in average must be delivered by cesarean section, but Gross et al. [7] considered 5-cm to be the critical limit. We consider that every infant with a sacrococcygeal teratoma, without associated anomalies incompatible with life, must be delivered by cesarean section, since the average size of the tumor is added to the normal dimensions of the fetal girdle. An absolutely atraumatic vaginal delivery in these cases is therefore not warranted. Using either the classical cesarean section [17] or the low-segment cesarean section with an inverted T-shaped hysterotomy provide the least traumatic approaches to the fetus. Since these tumors are potentially malignant and the incidence of malignancy increases with age, complete resection of the tumor with excision of the coccys in the first week of life and pathologic examination of the resected mass are mandatory [5, 16, 18].

This case report agrees with previous reported cases in which successful management of sacrococcygeal teratomas could be achieved with accurate prenatal sonographic diagnosis, atraumatic delivery, and prompt surgical intervention [2, 7, 9, 17].

Abstract

Teratomas are tumors containing tissues derived from all three germinal cell layers. Most teratomas arise in the sacrococcygeal region, with a reported incidence of 1/40.000 births. During labor, severe dystocia may occur in infants with large tumors if delivered vaginally, with an extremely high fetal morbidity and mortality rate. Reported is a case of a large congenital sacrococcygeal teratoma diagnosed antepartum by sonography at 36 weeks pregnancy. This sonographic finding allowed us to avoid traumatic delivery. Cesarean section was performed, with the delivery of a 4.050-g female infant with a large sacrococcygeal tumor. Resection of the sacral mass was performed on the 5th day of life. Pathologic examination showed a mature sacrococcygeal teratoma weighing 620-g, and measuring 14-cm by 11-cm by 9-cm. The infant had an uneventful recovery.
This report agrees with previous reported cases in which successful management of sacrococcygeal teratomas could be achieved with accurate prenatal diagnosis, atraumatic delivery, and prompt surgical intervention. The role of sonography in the management of these cases is stressed.

Keywords: Fetal disease, fetal malformation, prenatal diagnosis, sacrococcygeal region, teratomas, ultrasound studies.

Zusammenfassung

Pränatale sonographische Diagnose von kongenitalen Teratomen im Kreuz- und Steißbeinbereich und weiteres Vorgehen

Teratome sind Tumoren, die von allen drei Keimblättern abstammendes Gewebe enthalten. Die meisten Teratome wachsen in der Kreuz- und Steißbeinregion, wobei eine Inzidenz von 1/40 000 Geburten angegeben wird. Sub partu können schwere Dystokien auftreten, wenn Kinder mit großen Tumoren vaginal entbunden werden, wobei die fetale Morbidität und Mortalität extrem hoch ist. Wir berichten über einen Fall mit einem großen Teratom im Kreuz- und Steißbeinbereich, wo die Diagnose sonographisch in der 36. Schwangerschaftswoche gestellt wurde. Damit wurde eine traumatische Entbindung verhindert und eine Sectio durchgeführt. Das weibliche Neugeborene wog 4050 g und hatte einen großen Tumor in der Kreuz- und Steißbeinregion. Die Resektion des Tumors wurde am 5. Lebenstag durchgeführt. Die pathologische Untersuchung bewies ein reifes Teratom der Kreuzbein-/Steißbeinregion mit einem Gewicht von

620 g und einer Größe von 14 × 11 × 9 cm. Die Heilung bei dem Kind verlief komplikationslos.
Dieser Fall bestätigt vorangegangene Berichte, in denen beschrieben wird, daß eine exakte pränatale Diagnose, eine nicht traumatisierende Entbindung und eine sofortige chirurgische Intervention ein erfolgreiches Management bei Vorliegen von Teratomen im Kreuz- und Steißbeinbereich ermöglichen. Von besonderer Bedeutung in diesen Fällen ist die Sonographie.

Schlüsselwörter: Fetale Erkrankung, fetale Mißbildung, Kreuzbein-/Steißbeinregion, pränatale Diagnose, Teratome, Ultraschalluntersuchung.

Résumé

Diagnostic échographique prénatal d'un tératome congénital sacrococcygien et prise en charge
Les tératomes sont des tumeurs constituées de tissus provenant des trois feuillets germinaux. La plupart des tératomes diègent dasn la région sacrococcygienne avec une incidence de 1/40 000 naissances. Une dystocie grave peut apparaitre en cours de travail chez les enfants qui présentent des dumeurs volumineuses, si l'accouchement se fait par voie basse, avec une morbidité et mortalité extrêmement élevées. On rapporte un cas de tératome sacrococcygien congénital volumineux, diagnostiqué avant l'accouchement par échographie à 36 semaines de grossesse. Les images échographiques nous ont permis d'éviter un accouchement traumatique. Une césarienne a été effectuée, qui a donné naissance à une fille de 4050 g porteuse d'une volumineuse tumeur sacrococcygienne. L'exérèse de la masse sacrée a été pratiquée au 5ème jour de vie. L'examen anatomopathologique a mis en évidence un tératome sacrococcygien de 620 g, mesurant 14 cm sur 11 cm sur 9 cm. Les suites ont été simples pour l'enfant. Cette observation s'accorde avec les cas précédents rapportés pour lesquels une prise en charge satisfaisante a pu être réalisée grâce à un diagnostic prénatal approprié, un accouchement atraumatique et une intervention chirurgicale rapide.
On insiste sur le rôle de l'échographie dans la prise en charge de tels cas.

Mots-clés: Diagnostic prénatal, études échographiques, Maladie fœtale, malformation fœtale, région sacrococcygienne, tératomes.

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